

Thyroglossal duct cyst in hyoid bone: Unusual location

S. S. Bist · Manisha Bisht · Saurabh Varshney · Nitin Gupta · Rajat Bhatia

Abstract

A 10-year-old female presenting with a discharging sinus in the midline of the neck of one month duration was diagnosed to have a thyroglossal sinus. She underwent sistrunk's operation, intra-operatively a thyroglossal duct cyst was atypically sited in the intrahyoid region. The cyst may be located in the intralingual, suprathyroid, thyrohyoid or suprasternal region. A literature search revealed that this intrahyoid location of a thyroglossal duct cyst is extremely rare and so far only three cases have been reported. A case and review of literature regarding this unusual entity are presented.

Keywords Thyroglossal Duct Cyst · Thyroglossal Cyst · Hyoid Bone

Introduction

Thyroglossal duct cyst is the most common congenital neck mass, resulting from the persistence and dilatation of remnants of an epithelial tract formed during migration of the thyroid during embryogenesis. Approximately 7 per cent of the population have thyroglossal duct remnants and it accounts for 70 per cent of congenital neck abnormalities [1]. A thyroglossal duct cyst or remnants presents in childhood before the age of six in 76 per cent of cases [2]. Thyroglossal duct cyst commonly presents as painless midline neck swelling [1, 2] and is most frequently found below the level of the hyoid bone (85%). But they may be situated anywhere from the region of the foramen caecum at the base of the tongue to the level of suprasternal notch. They may also be found lateral to the midline, mainly on the left, in 10–20 per cent of the cases, probably because of the levator glandulae thyroideae muscle on that side. Classically the cyst moves upward on protruding the tongue (because of its attachment to the base of the tongue via the thyroglossal tract) and this physical sign is virtually pathognomonic of it. A thyroglossal duct cyst occurs due to the failure of obliteration of the thyroglossal duct. However, thyroglossal sinus is always acquired, secondary to spontaneous or surgical drainage after infection. Here we report a young female child with thyroglossal sinus secondary to incision and drainage along with thyroglossal duct cyst located with in hyoid bone.

Case History

A 10-year-old female presented with a history of midline neck swelling in the upper part of the neck, insidious in onset and of one-year duration. The swelling had rapidly increased in size over the past one month, for which she consulted a local doctor who had performed incision and drainage for the same. The patient then presented in our hospital with a discharging sinus in the upper part of the neck in the midline (Fig. 1). On examination, a small swelling

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was located in the anterior neck, at the level of hyoid bone along with a sinus opening in the thyrohyoid region. This swelling along with the sinus was mobile with protrusion of the tongue. There was associated tender cervical lymphadenopathy in submental region, due to infection. Laboratory tests were normal and ultrasound neck confirmed thyroid gland in normal position. Sinogram revealed the sinus tract extending up to the hyoid bone. Subsequently, the patient was planned for sistrunk's operation under general anesthesia. Intraoperatively it was found that the tract was passing through the body of hyoid bone and the cyst had splayed out mid portion of the hyoid bone. A sistrunk's procedure was performed wherein the sinus tract, cyst, the body of hyoid bone, a cuff of central base tongue extending superiorly and inferiorly extending tract were all removed in toto (Fig. 2). Postoperative recovery was uneventful. No recurrence of the disease has been seen for the past one year. Histological examination of the specimen shows a sinus tract with chronic inflammation consistent with thyroglossal cyst related sinus tract.

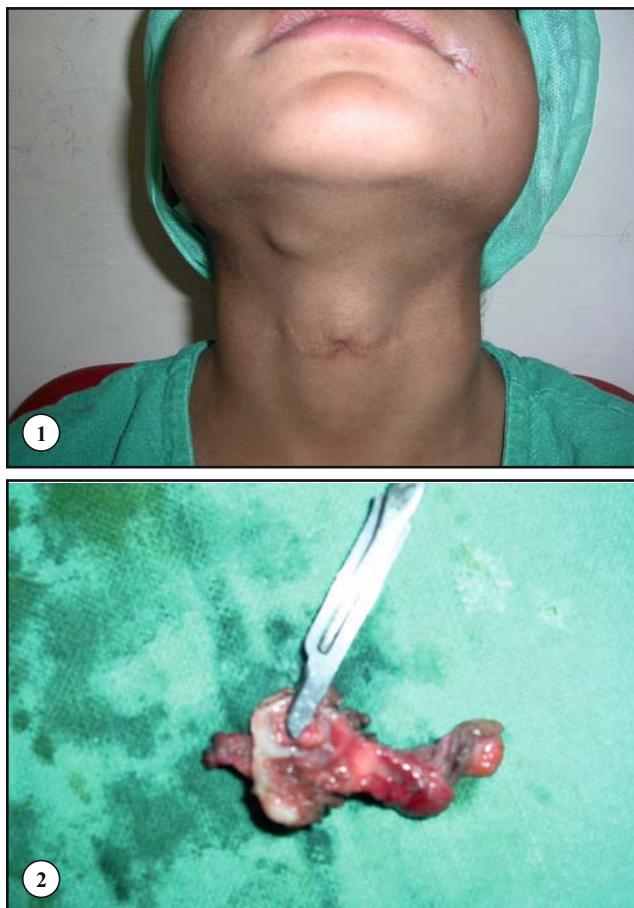


Fig. 1 Clinical photograph showing thyroglossal sinus with cervical lymphadenopathy

Fig. 2 Photo showing the excised specimen of the thyroglossal duct sinus with cyst, including the splayed out body of the hyoid bone.

Discussion

Although thyroglossal duct cysts generally presents clinically in children, it is important to understand that the lesion can present in adults as well, sometimes much later in life [1]. The thyroid gland develops during the fourth week of intrauterine life as a median thickening in the ectoderm of the floor of the pharynx between the first and second pharyngeal pouches. The epithelial proliferation invaginates to form a thyroid diverticulum, which descends in the anterior neck to reach the adult site by the seventh week. The thyroid gland remains connected to the foramen caecum by a thyroglossal duct, which eventually involutes and disappears. A thyroglossal duct cyst may appear at any point in the migratory pathway taken by the thyroid gland during its development including the thyroid gland per se. There are four general location of the thyroglossal duct cyst – intralingual (2.1%), suprathyroid (24.1%), thyrohyoid (60.9%) and suprasternal (12.9%) [3]. In 1940, Frazer [4] discussed the intimate relationship between the tract and the hyoid bone. He noted that the hyoid bone changes from ovoid shape in the embryo to a crescentric shape in the adult. In the adult, therefore, persisting thyroglossal tract will pass down in front of the hyoid bone and then hook up around its inferior border to lie posterior to the bone before finally descending to the isthmus. However several authors have stated that the tract may descend posterior to or even through the hyoid bone [5]. The forward growth of the hyoid bone explains the siphon like widening of the thyroglossal tract around the lower and posterior surfaces of the body of the hyoid bone. According to other theories, however, the embryonic thyroid can develop along a pre, trans, or retrohyoid pathway. The intrathyroid location is explained by one of the theories of the migration of the rudimentary thyroid during embryogenesis. A retrohyoid and especially intrathyroid localization is rare and is usually considered as a simple cyst attached to the periosteum [6]. In the present case, the thyroglossal duct cyst was located within the hyoid bone and cyst splayed out the hyoid bone. This feature has been reported in three case reports in the literature [5, 6, 7]. Horiseva et al [8] reported one case in 59-year-old man in which the thyroglossal duct penetrated the hyoid bone. Sistrunk [9] described the thyroglossal ducts as usually passing through the hyoid bone, although they are sometimes found to pass anteriorly or posteriorly to the hyoid bone. This is uncommon as borne out by large series reported [2]. Sistrunk's procedure first described in 1920, involves two essential elements: 1) excision of a core of tissue from hyoid to foramen caecum with the deliberate aim of staying within normal tissue 2) removal of central portion of the hyoid bone. A well-performed “classical” sistrunk operation has a success rate of 95–97 per cent. Ellis and Van Noststrand [10] in 1977 reintroduced the theory that thyroglossal duct passes anteriorly to the hyoid bone. Thyroid tissue is present in thyroglossal duct cyst wall in more than 60% of cases [3]. In our case, inactive thyroid tissue was observed.

It is now recommended that in all suprathyroid and infrathyroid swelling consideration should be given to order ^{131}I scan to locate the functioning thyroid. Although in former years there was not the same necessity to scan patients with lumps in the usual position between the thyroid and front of the hyoid the medico-legal position now dictates that all should have a thyroid scan. Our patient could not undergo the same due to financial constraints, so only the presence of thyroid gland in normal position was confirmed by ultrasound neck.

Conclusion

The thyroglossal duct varies in its relationship to the hyoid bone so that it may not only be anterior or posterior but may rarely present through the substance of the hyoid bone. However, it is well established that the tract is intimately related to the hyoid bone and the attempts to dissect the tract from its surface are likely to fail. The best way to remove all tract remnants and thus avoid recurrence is to excise the central part i.e. the body of the hyoid bone. This intrathyroid localization supports the surgical approach of systematic resection of the body of the hyoid bone in thyroglossal duct cyst cases.

- This is a case report of an atypical thyroglossal cyst arising within the hyoid bone
- Three such cases have been previously reported
- The embryology of this finding is briefly discussed

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